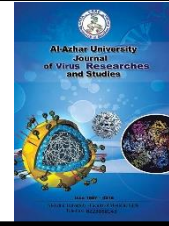




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### Different Modalities in Management of Congenital Clapsed Thumb

Tamer Abd Elkader Abd Elwahed<sup>1</sup>, Mahmoud Ali Ismail<sup>1</sup>, Ashraf Abdelaziz<sup>1</sup>,  
Ehab Abd AL Fattah Abd Allah<sup>1</sup> and Tharwat Mohamed Abd Alrahman<sup>2</sup>

<sup>1</sup>Department of Orthopedic Surgery, Faculty of Medicine, Al-Azhar University, Assiut, Egypt.

<sup>1</sup>Department of Orthopedic and Traumatology, Faculty of Medicine for Girls, Al-Azhar University.

\*E-mail: TamerAbdElkader.216@azhar.edu.eg

#### Abstract

This study aimed to evaluate the management of patients with congenital clasped thumb deformity. This is a prospective cohort study, conducted in multicenter of Al-Azhar University Hospitals. It included 20 pediatric patients below eight years old with congenital clasped thumbs. All the patients' demographics (age, and sex) and clinical data regarding the presence of other associated anomalies such as congenital talipes equinovarus (C.T.E.V.) and arthrogyposis were recorded. All the patients were exposed to thorough clinical examinations (performed by a pediatrician). The studied patients were classified according to Tsuyuguchi et al. [1] classification into three types. Conservative treatment was sufficient in one case, and surgical treatment was performed for 18 patients using different types of flaps (Elghani flap in 85%, and Ezaki flap in two cases (10.0%)) with an average follow-up of 20 months from the start of January 2019 up to the end of August 2020. Twenty patients were included with a mean age of  $5.65 \pm 1.84$  years and ranged from 2 up to 8 years, 12 (60%) were boys versus 8 (40.0%) were girls. Positive family history was recorded in 20.0% of studied cases. Associated anomalies were reported in 45.0% of our cases. Almost all patients were satisfied with the surgical treatment result. Early proper management of congenital clasped thumb deformity results in good satisfactory outcomes.

**Keywords:** Thumb, Congenital clasped thumb, Management.

#### 1. Introduction

A persistent progressive flexion and adduction of the thumb after the 3rd or 4th month of life is called congenital clasped thumb (CCT).[2] It is a rare deformity presenting with heterogenous congenital abnormalities, [3, 4] mostly confused with trigger finger deformity due to fixated flexion deformity of the thumb.

Entrapment of the trigger finger is found at the A1 pulley level in congenital trigger finger due to fusiform hypertrophy of the flexor pollicis longus tendon. [3, 5, 6] The fundamental issue is the deficient extensor tendon mechanism in the thumb, which is caused by both structural and functional factors. [3, 5, 7, 8]

Although in CCT there is thumb flexion deformity due to extensor tendon insufficiency and skin and muscle contractions in the flexor and thenar areas. [3, 8, 9]

Success of CCT treatment depends on type of the lesion. Although successful results have been obtained with splinting and conservative treatment, several reconstruction methods are used due to secondarily developed contractures in neglected cases. Tendon transfers are useful in hypermobile thumb cases who have both extensor and opponents tendon deficiency. [3, 10]

So, this study was aimed to evaluate the management of congenital clasped thumb deformity in pediatric patients.

## 2. Patients and Methods

Prospective cohort study registered at Clinical trial. That was conducted at Al-Azhar University Hospitals from the start of 2019 up to the end of 2020. The protocol of the study was approved by Dean of Faculty of Medicine, Azhar Assiut and Azhar Cairo for Girls University Medical Ethical Review Board.

### 2.1 Eligible participants

We included all patients less than eight years old, with congenital clasped thumbs who were in need of surgical correction and rehabilitation. The exclusion criteria patients more than eight years old, with congenital trigger thumb, neglected adducted thumb, post-traumatic extensor tendon injury, neglected congenital clasped thumb and patients whose parents refused to participate in the study were also excluded. Written consent was obtained from all parents of the participants of the study. Then, patients were subjected to detailed history including demographic data and full clinical examination. According to Tsuyuguchi et al. [1] deformed thumbs were classified into:

**Type I:** supple clasped thumb. The thumb could be passively abducted and extended against the resistance of thumb flexors, without other digital anomalies.

**Type II:** clasped thumb with hand contractures. The thumb could not be passively abducted and extended, with or without other fingers anomalies.

**Type III:** clasped thumb associated with arthrogyposis.

### 2.2 Treatment protocol

- *Non-operative treatment in type 1:* full-time splinting of the thumb in extension for at least 6 months, followed by night splinting for further 6 months after achieving active extension of the thumb.
- *Operative treatment in type 2 and type 3:* surgical procedures for every case were variable according to the degree of narrowing of the first web, stability of the metacarpophalangeal (MP) joint and muscle deficiency. [11]

The surgery aimed to widen the narrow first web space, including both the skin and deep tight structures. Skin widening was done through either Ghani flap [12] in 85.0% or Ezaki flap [13] in 10.0%. The used technique was variable according to the degree of narrowing.

The first metacarpus is kept in full abduction after full release by crossing two k-wires over the first web gap. Chondrodesis was required in situations of severely unstable MP joints, however double breasting of the capsule was done in cases of modest ulnar collateral ligament laxity. In both cases, the MP joint was fixated with a single or two crossed k-wires. When the MP joint was stable or after obtaining stability with ulnar collateral ligament restoration, tendon transfer was performed to restore active extension. The extensor indicis was the preferred tendon for transfer, but if it was unavailable, one slip of the flexor digitorum superficialis muscle was transplanted to the deficient thumb

extensors' vestigial remnant. Lengthening of the flexor pollicis longus (FPL) was done after the release of the palmar contracture of the thumb, it was needed in a few cases because in the majority of cases we did chondrodesis of the MP joint. Z-lengthening or intramuscular tenotomy is used to lengthen the distal forearm.

### 2.3 Rehabilitation

An above-elbow splint was applied immediately. After 6 weeks of surgery, the k-wires were removed. For at least 6 months after surgery, the position was kept in a night splint with the thumb extended, with daily active use of the thumb being recommended.

### 2.4 Follow up

All cases will be followed every 2 weeks in the first 2 months then every month for 6 months then every 2 months for one year. The average duration of follow-up was about 20 months. Due to the difficulty in testing thumb function at that young age, there were no common criteria for evaluating the results of therapy of clasped thumb [11]. To assess the thumb function, we assessed; the parent's satisfaction, cosmetic appearance, stability of the first MP joint, and the thumb functions (abduction, extension and rotation).

### 2.5 Statistical Analysis

All statistical calculations were done using SPSS (statistical package for the social science; SPSS Inc., Chicago, IL, USA) version 22. Data were described in the form of mean  $\pm$  standard deviation or number (percent) as appropriate.

## 3. Results

The mean age of studied participants was  $5.65 \pm 1.84$  years with a range (of 2 to 8 years old), 12/20 (60.0%) were boys versus 8/20 (40.0%) were girls. For laterality, 13

(65.0%) with right-side clasped thumb (CT), 6 (30.0%) with left-side CT and one case with bilateral CT, positive family history was documented in four cases (20.0%). Forty-five percent suffered from associated congenital anomalies (25.0% suffered from Congenital talipes equinovarus (C.T.E.V) and 20.0% suffered from arthrogryposis), and no cases suffered from associated syndromes. Twenty percent have complex CT and 80.0% have simple CT (Table 1).

The operative findings revealed that, the extensor tendons were absent in 45.0% and attenuated in 55.0%, widening of first web space was done in 95.0% of studied cases, and only one case (5.0%) have no such contracture and have received just stretching of the first web space. Fractional lengthening was done for six studied cases (30.0%), meanwhile for the majority of studied cases (65.0%) we did chondrodesis of the MP joint. Thirteen (65.0%) received extensor indices transfer in, two cases (10.0%) received ring finger flexor digitorum superficialis (FDS), and the remaining 10.0% received augmentation of extensor pollicis longus muscle (EPL), and 15% don't receive tendon transfer. Elghani flap was used in most of our studied cases 17/20 (85.0%), and Ezaki flap was used in the remaining two cases (10%) (Table 2).

Post-operative evaluation of the thumb functions revealed that; regarding the degree of parent's satisfaction and the degree of hand grip, 50% reported excellent satisfaction and excellent degree of hand grip, 30% reported good satisfaction and good degree of hand grip, 10% reported fair satisfaction and fair degree of hand grip and 10% reported poor satisfaction and poor degree of hand grip, for the thumb functions; 50% of studied cases had excellent abduction and opposition, and 30% thumbs had good abduction and opposition. No case had excellent thumb rotation, and 80% had good thumb rotation, MP joint was unstable in eleven operated hands (55%). Regarding complications,

two hands (10%) with non-union of the chondrodesis, revision of chondrodesis was

needed in both of them because both thumbs have poor stability (Table 3).

**Table (1):** Distribution of the cases according to age (n=20).

	N	(%)
<b>Age (years)</b>		
• Range	2 - 8	
• Mean $\pm$ SD	5.65 $\pm$ 1.84	
<b>Sex</b>		
• Male	12	(60.0)
• Female	8	(40.0)
<b>Side</b>		
• Right	13	(65.0)
• Left	6	(30.0)
• Bilateral	1	(5.0)
<b>Family history</b>		
• Positive	4	(20.0)
• Negative	16	(80.0)
<b>Associated anomalies</b>		
• No	11	(55.0)
• C.T.E.V.	5	(25.0)
• ARTHROGRYPOSIS	4	(20.0)
<b>Associated syndromes</b>		
• Yes	0	(0.0)
• No	20	(100.0)
<b>Types of clasped thumb</b>		
• Complex	4	(20.0)
• Supple	16	(80.0)

Quantitative data are presented as mean  $\pm$  SD and range; qualitative data are presented as number (percentage).

**Table (2):** The operative findings among the studied cases (n=20).

	N	(%)
<b>Extensor Tendons</b>		
• Absent	9	(45.0)
• Attenuated	11	(55.0)
<b>First web space contracture</b>		
• Stretching	1	(5.0)
• Widening	19	(95.0)
<b>Flexor tendons</b>		
• Fractional lengthening	6	(30.0)
• No	14	(70.0)
<b>Chondrodesis CMPC or K-wire</b>		
• Yes	13	(65.0)
• No	7	(35.0)
<b>Tendon transfer</b>		
• Augmentation of EPL	2	(10.0)
• E.I. transfer	13	(65.0)
• FDS of ring finger	2	(10.0)
• No	3	(15.0)
<b>Types of flaps</b>		
• Elghani flap	17	(85.0)
• Ezaki flap	2	(10.0)
• No flap used "just stretching"	1	(5.0)

Qualitative data are presented as number (percentage).

**Table (3):** Post-operative evaluation of the thumb function among the studied cases (n=20).

	N	(%)
<b>Parent's satisfaction</b>		
• Excellent	10	(50.0)
• Good	6	(30.0)
• Fair	2	(10.0)
• Poor	2	(10.0)
<b>Hand grip</b>		
• Excellent	10	(50.0)
• Good	6	(30.0)
• Fair	2	(10.0)
• Poor	2	(10.0)
<b>Abduction</b>		
• Excellent	10	(50.0)
• Good	6	(30.0)
• Fair	2	(10.0)
• Poor	2	(10.0)
- Range	10 - 45	
- Mean $\pm$ SD	33.5 $\pm$ 10.53	
<b>Rotation</b>		
• Excellent	0	(0.0)
• Good	16	(80.0)
• Fair	2	(10.0)
• Poor	2	(10.0)
- Range	70 - 100	
- Mean $\pm$ SD	92.00 $\pm$ 10.05	
<b>Opposition</b>		
• With little (excellent)	10	(50.0)
• With middle (fair)	4	(20.0)
• With ring (good)	6	(30.0)
• None	0	(0.0)
<b>MCP</b>		
• Stable	9	(45.0)
• Unstable	11	(55.0)
<b>Post-operative complication</b>		
• Revision	2	(10.0)
• No	18	(90.0)

Quantitative data are presented as mean  $\pm$  SD and range; qualitative data are presented as number (percentage)

#### 4. Discussion

Congenital clasped thumb (CCT) is a progressive flexion and adduction deformity presenting with heterogeneous congenital abnormalities and syndromes. This deformity is frequently accompanied by the narrowing of the first web space and laxity of the metacarpophalangeal joint (MPJ). Treatment requires a thorough understanding of the clasped thumb's varied characteristics and an accurate diagnosis. Treatment options range from conservative to surgical, depending on the classification [14]. The present work demonstrates better outcome among studied cases with better quality of life and high patient parent's satisfaction.

The congenital clasped thumb was diagnosed by persistent flexed adducted thumb after the 3rd or 4th month of life, and it is difficult to diagnose congenital clasped thumb before that time as it is normal that the thumb to be clutched into the palm in these first months beside that the disease is rare [3]. For this reason, we include children aged from 2 up to 8 years old with a mean age of  $5.65 \pm 1.84$  years. In contrast the prospective study of Ghani et al., 2007 [11] reported that the median age of studied participants was 27 months and ranged from 3 months to 11.5 years. This difference is attributed to different inclusion criteria.

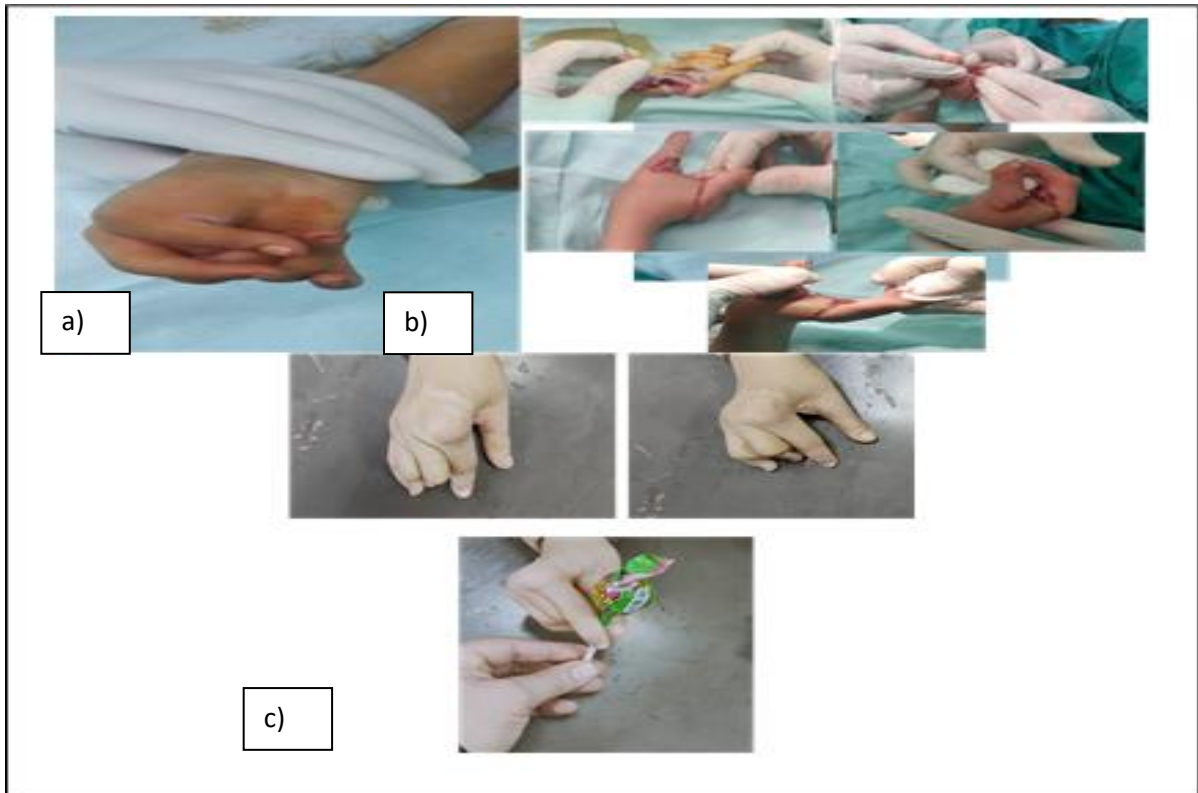
Most of previous literature has discussed surgical techniques or case reports of associated anomalies or syndromes. Little is known about the prevalence and epidemiology of CT. Furthermore, there is a lack of information in the literature about the characteristics of patients with CT.

Out of 20 patients with CCT, 12 (60%) were boys versus 8 (40.0%) were girls with male: female ratio 1.5:1. This figure is in line with the previous studies of WECKESSER et al., 1968 [15], Flatt, 1994 [16], Lin et al., 1999 [10], Ghani et al., 2007 [11] as all reported that congenital clasped thumb occurs more often in males

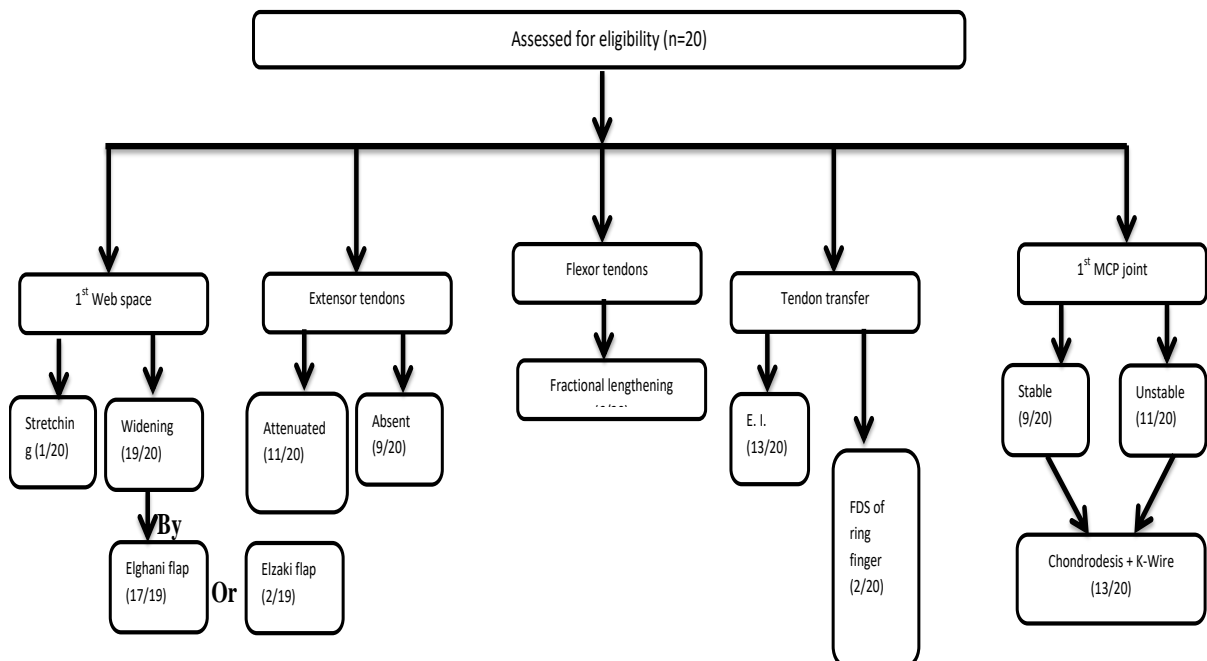
than in females. However, it differs from the 1:1 ratio reported by Tsuyuguchi et al., 1985 [1]. The wide variety of syndromes associated with CCT and their various modes of inheritance, as well as the minimal number of studies in the literature that describe the sex ratio of affected patients, can explain this disparity.

Regarding lesion laterality, 13 (65.0%) with right side, 6 (30.0%) with left side and only one case (5.0%) suffered from bilateral clasped thumb. Conversely the previous studies of WECKESSER et al., 1968 [15], Flatt, 1994 [16], Lin et al., 1999 [10], Ghani et al., 2007 [11] reported bilaterally in > 80 %.

Regarding possible predisposing factors of CCT we found that 20.0% have a positive family history. Our study is near to the estimated percentage reported by WECKESSER et al., 1968 [15] and Ghani et al., 2007 [11] who reported positive family history of 32–36 %. On the other hand Tsuyuguchi et al., 1985 [1] reported that only 10% have positive family history of CCT. The high rate of bilateral deformities suggests that a defect exists in the zygote prior to cell division, also the familial occurrence of this anomaly adds to the evidence for genetic defect [17]. This raises our awareness for the need of genetic assessment of all cases with CCT by a pediatric geneticist to determine and control possible predisposing factors for such condition. Most of the cases of CCT are part of generalized disorders. Our study confirms the previously reported high incidence of associated congenital anomalies with CT [1, 2, 6, 18-27] as 45.0% of our patients were found to have associated anomalies; 25.0% suffered from C.T.E.V and 20.0% suffered from arthrogryposis. Because the different syndromes were investigated separately or the literature contains case reports of particular syndromes, the percentage of related congenital abnormalities with clasped thumb varies depending on the number of syndromes included in the literature.



**Figure 1:** a) **Pre-operative:** right hand clapsed thumb associated with arthrogryptic anomalies and flexion contracture of the other fingers. b) **Intra-operative:** widening of first web space, chondrodesis of first MCP joint and k-wire fixation using Elghani flap. c) **Post-operative:** follow up with a good position of the thumb “out of the palm” and a good hand grip.



**Figure 2:** Flow diagram showing the sequence of the procedure done for the studied clapsed thumb cases.

In line with our study Temtamy, 1978 [2] reported that 15% have upper limb anomalies and Ghani et al., 2007 [11] reported that 77.5% of studied cases have congenital anomalies with CT, 15% have upper limb anomalies, 25% have arthrogryposis, other anomalies encountered included congenital heart disease in the form of ventricular septal defect, which was detected in one patient who had no other anomalies except unilateral CT, two patients with congenital blindness in association with bilateral CCT, and a case with radial deviation and rotation of the index finger.

No abdominal anomalies were encountered in this study, and this agrees with the previous studies of [2, 11, 28]

Our study revealed that no cases suffered from associated syndromes. On the other hand Ghani et al., 2007 [11] reported that 68% of studied patients have associated syndromes.

In 1968 Weckesser et al [15] proposed a staging system which classified patients with CCT into 4 groups according to the severity of deformity. In 1985 Tsuyuguchi et al. [1] classified patients with CCT into three groups. Ghani et al. (2007) [11] found no difference between type II and type III clasped thumbs in terms of pathological findings, severity, treatment protocol, operative procedures, and results; thus, the author concluded that classifying the clasped thumb into supple and complex types, as McCarroll [29] did, was a better idea. In our study we used the classification suggested by McCarroll Jr, 1985 [29], and we found that 20.0% of studied cases have complex CT and 80.0% have supple CT.

Treatment for a CCT is dependent on the stage of the disease, the age at presentation, and any associated pathologies [3]. Conservative treatment was effective for patients under 1 year of age, according to Ghani et al., 2007 [11] while surgery was preferable for type II and type III patients who did not respond to conservative treatment. For patients who presented beyond 2 years of age and do not respond

to conservative treatment, Medina et al., 2008 [5] suggested a surgical procedure. McCarroll Jr, 1985 [29] documented that splinting can help with the flexible type, but when skin contracture and/or ligament laxity are present, surgery is required.

Our protocol of management is similar to that followed by McCarroll [29], Lipskeir and Weizenbluth, 1989 [30] and Ghani et al., 2007 [11] as follow; the flexible form was treated with splinting and, if splinting failed, tendon transfer, whereas the complex type was treated with correction of fixed contractures and restoration of lax ligaments and tight skin as needed. Meanwhile, Tsuyuguchi et al., 1985 [1] cases were treated by splinting for types I and II cases and operative treatment for type III and type II cases which didn't respond to conservative management.

The operative findings of our study revealed that the extensor tendons were absent in 45.0% and attenuated in 55.0%. On the other hand Crawford et al., 1966 [31] and McCarroll Jr, 1985 [29] reported that the extensor tendons were not absent but attenuated. And Ghani et al., 2007 [11] reported that attenuated extensor pollicis longus and brevis were found in all hands. Absent extensor indicis was detected in one out of eight hands with tendon transfer.

Also, our study shows that 95.0% of studied cases have contractures in the soft tissues in the volar surface of the digit, involving all tissues including skin, subcutaneous fascia, periarticular structures and in the radial collateral ligament–volar plate complex of the MP joint and only one case (5.0%) have no such contracture and have received just stretching of the first web space. Global instability of the MP joint was present in 11 hands (55%). Meanwhile; Ghani et al., 2007 [11] reported instability of the MP joint in 18/73 hands.

The shortening of the flexor pollicis longus muscle was present in 6 (30.0%) of studied cases and need fractional lengthening because in the majority of cases (65.0%) we did chondrodesis of the MP joint.



Tendon transfer was done to restore active extension of the MP joint when it was stable or after achieving stability with reconstruction of the ulnar collateral ligament [17]. The extensor indices were the preferred tendon for transfer [32] it was done in 65.0% of studied cases, or the ring finger FDS if extensor indices were absent [33], it was used in 10.0% and transferred to the vestigial remnant of the deficient thumb extensors, and the remaining 10.0% have received augmentation of EPL; 15% don't receive tendon transfer.

To increase the web space in our study, we performed several skin incisions. Because web release relies on the severity of web narrowing and degree of contracture, no methodology can be standardized; as a result, comparing the various procedures utilized for release is challenging.

To evaluate the thumb in our study function we used the same combination as described by Ghani et al., 2007 [11]. The author used a combination of criteria because it is of no benefit to achieve active thumb extension without achieving stability of the MP joint or without widening of the web. It is very important to consider the stability of the MP joint as follows: parent's satisfaction, cosmetic appearance, thumb functions (Abduction, extension and rotation) and the degree of stability of the first MP joint. Results of the present study reported that, for parent's satisfaction and hand grip, 50% reported excellent satisfaction and excellent degree of hand grip, 30% reported good satisfaction and good degree of hand grip, 10% reported fair satisfaction and fair degree of hand grip and 10% reported poor satisfaction and poor degree of hand grip. Similarly; Ghani et al., 2007 [11] reported that the parents of all the patients were satisfied with the results.

For thumb functions, 50% of studied cases had excellent abduction and opposition, and in 30% thumbs had good abduction and opposition. No case had excellent thumb rotation, and 80% had good thumb rotation. This figure is better than Ghani et al. [11] study who reported that 24/73 thumbs had

excellent abduction, and 4/73 thumbs had good abduction. 26/73 thumbs had excellent rotation, and 2/73 had good rotation.

For MP joint stability; it was unstable in 11 hands (55%) of our studied cases, on the other hand Ghani et al., 2007 [11] reported that the stability of the MP joint of the thumbs was excellent in 24/73 cases (60%). As regards the post-treatment complications, two hands (10%) with non-union of the chondrodesis, revision of chondrodesis was needed in both of them because both thumbs have poor stability. In line with our study Ghani et al., 2007 [11] reported that the two hands with non-union of the chondrodesis, revision of chondrodesis was needed in only one thumb with poor stability, while in the other the stability was fair and there was no need for surgery; otherwise, the rest of cases with chondrodesis had excellent stability.

Also our study is not without limitation; as the degree of consanguinity which is one of the possible predisposing factors for CCT was not reported in our study, genetic assessment of CCT studied cases were not done, cosmetic appearance of the hand was not measured in our study, small sample size, and also time required to reach the satisfied level of thumb function was not reported in our study.

## 5. Conclusion

Based on our finding we could concluded that; management of CCT by splinting with physical treatment is a very successful method in flexible cases or in patients who only have extensor tendon weakness, also properly planned treatment according to the type of the deformity improves the cosmetic appearance and functional capabilities of the hand. Both used flaps are associated with the same treatment outcomes.

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